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Dental Anomalies among Down Syndrome Children in Khartoum State (Sudan)

By:

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Declaration

I declare that this work is original and that it is as partial fulfillment of MSc. degree in Pediatric Dentistry according to the Guidelines of the University of Khartoum Graduate Collage of Medical and Health Sciences Board. This work has not been submitted elsewhere.

Dedication

To my lovely parents Hassan & Fathia

To My sisters & brothers

and

To all whom I love

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Acknowledgment

First of all I would like to thank Allah the Almighty, the Merciful, who gave me the power and health to finish this work.

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Abbreviations

DPTs	Dental panoramic tomographies
DS	Down syndrome
ICD	International classification of diseases
IQ	Intelligence Quotient
SD	Standard deviation
U Of K	University of Khartoum

Abstract

Background: Down syndrome (DS) is a genetic alteration in which the affected individual instead of 46 has 47 chromosomes, carrying an extra pair in chromosome # 21. Dental anomalies such as missing/extra teeth, abnormal tooth shape/size and delayed eruption of the permanent teeth are reported to be common in DS.

Design: Descriptive, cross-sectional, institution-based study.

Setting: Rehabilitation Centers for Children with Special Needs in Khartoum State and the Clinic of Pediatric Dentistry, University of Khartoum.

Objective: This study aimed to assess the dental anomalies among a group of institutionalized Sudanese children with DS aged 6 to 14 years.

Methods: Ninety two DS children were included in this study, their mean age was 10.4 (± 2.8). Dental anomalies were evaluated by clinical dental examination and interpretation of panoramic radiographs. The participants were examined for the presence or absence of anomalies such as missing/extra teeth, abnormal tooth shape/size and delayed eruption of the permanent teeth.

Results: Children with dental anomalies represented 55.4% of the sample, while those without were 44.6%. Among those with anomalies 51% were males and 49% were females. Most of the cases (66.7%) had one type of anomaly; among which hypodontia was the most common type (54.9%) and it was found to be more in the mandible than in the maxilla. Taurodontism was the second anomaly representing 49%, followed by supernumerary teeth (17.6%). Other anomalies showed very low incidence.

Conclusion: The prevalence of dental anomalies in DS children found in the present study, is similar to those of the reviewed literature. Except for hypodontia and taurodontism, other anomalies showed a low incidence.

ملخص البحث:

خلفية: متلازمة داون هي مرض وراثي، الشخص المصاب به لديه 47 كروموسوم بدلا عن 46 أي أن لديه كروموسوم إضافي موجود بكروموسوم رقم 21 . الشذوذات التطورية السنية مثل نقص

وزيادة عدد الأسنان، إضطرابات حجم و شكل الأسنان و تأخربزوغ الأسنان الدائمة شائعة الحدوث لدى أطفال متلازمة داون.

تصميم البحث: دراسة مقطعية وصفية استنادا على معلومات مأخوذة من أطفال متلازمة داون الموجودين بمراكز ذوي الإحتياجات الخاصة.

مكان إجراء البحث: مراكز ذوي الأحتياجات الخاصة بولاية الخرطوم و عيادة طب أسنان الأطفال بكلية طب الأسنان جامعة الخرطوم.

الهدف من البحث : تهدف هذه الدراسة لتقييم أكثر أنواع الشذوذات التطورية السنية شيوعا لدى الأطفال السودانيين المصابين بمتلازمة داون الموجودين بمراكز ذوي الإحتياجات الخاصة للفئة العمرية 6- 14 سنة.

الأساليب : الحجم الكلي للعينة 92 طفل سوداني مصاب بمتلازمة داون متوسط أعمارهم 10.4 (± 2.8) سنة. الشذوذات التطورية للأسنان تم تقييمها بطريقتين: الفحص السريري بالعيادة و صور أشعة بانوراما. تم فحص الأطفال لتسجيل وجود الشذوذات التطورية أو عدمها مثل نقص

وزيادة عدد الأسنان، إضطرابات حجم و شكل الأسنان و تأخر بزوغ الأسنان الدائمة.

النتائج : الأطفال المصابين بالشذوذات التطورية السنية يمثلون 55.4 % من العينة أما الغير مصابين بها يمثلون 44.6%.

نسبة الأطفال المصابين بالشذوذات التطورية السنية كانت 51% ذكوراً و49% اناثاً. أغلب الحالات المصابة ظهر بها نوع واحد من الشذوذات (66.7%)، الغالبية العظمى منها كانت نقص عدد الأسنان (54.9%) ولوحظ أنها أكثر في الفك السفلي منه في العلوي. أما الإطالة في الغرفة اللبية (أسنان الثور) مثلت ثاني الأنواع شيوعا لدى هؤلاء الأطفال (49%) ثم الزيادة في عدد الأسنان (17.6%). أما باقي الشذوذات التطورية مثلت نسبة بسيطة جداً.

الخلاصة: معدل إنتشار الشذوذات التطورية بأسنان أطفال متلازمة داون في هذه الدراسة مشابه تماما لما هو موجود في الأدب الطبي.

خلافاً لنقص عدد الأسنان والإطالة في الغرفة اللبية (أسنان الثور) مثلت بقية الشذوذات التطورية السنية نسبة بسيطة جداً.

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|Chapter One

Introduction and Literature Review

1.1 Introduction

Down syndrome (DS) is an autosomal disorder in which the affected individual has 47 chromosomes instead of the normal 46 chromosomes, carrying an extra pair mate in chromosome # 21, thus known as trisomy 21. The disorder is characterized by generalized growth retardation and mental deficiency[1].

There are over 50 clinical signs of DS, but it is rare to find all or even most of them in one person. Some common characteristics include: midface dysplasia, flat and usually small nose associated with a low and broad nasal bridge, small maxilla, fissured tongue and papillary hypertrophy. Children with Down syndrome also have a smaller brain volume than other children, almond-shaped eyes, brachycephaly (a condition where the head is disproportionately wide), thick epicanthal folds with narrow palpebral tissue slanting towards the midline and poorly developed supra orbital ridge [2].

The teeth of cases with DS present complete mineralization. Tooth eruption may occur in an unusual order and can be 2 to 3 years behind a child's normal eruption pattern[3].

Over-retained primary teeth are also common in DS children. There is also a high incidence of impacted teeth [1].

Periodontitis is quite common in children with DS; however, a lower incidence of dental caries has been reported. The higher prevalence of periodontal disease in DS children is probably related to the impaired host response rather than to specific periodontal pathogens [4]. The low caries prevalence seems to be due to immune protection caused by the elevated salivary *S. mutans* specific IgA concentrations[5].

Malocclusion, particularly Angle's class III, is frequently seen due to the under-development of the midface [1].

Dental anomalies such as microdontia, macrodontia, missing & malformed teeth, small roots and taurodontism are also present with an incidence five times greater than in the normal population[6,7].

Early professional treatment together with daily care at home, can mitigate the severity of the outcome of dental anomalies in DS children, and allow them to enjoy the benefits of a healthy mouth [8].The main aim of this study is, therefore, to assess the most

common dental anomalies among a sample of institutionalized Sudanese children with DS.

1.2 Literature Review

1.2.1 Prevalence of Down syndrome:

Prevalence of (DS) is one in 700 live births and increases with increasing age of the mother at the time of conception[2].

1.2.2 Types of Down syndrome:

There are three types of DS [9]:

1.2.2.1 Trisomy 21:

Trisomy 21 is the most frequently occurring type of DS (approximately 95%). In this type, the individual with Down syndrome has 47 chromosomes in each cell instead of the usual 46, with 3 copies of the chromosome 21 instead of the usual pairTaurodontism came as the second most common anomaly, followed by supernumerary teeth. It was found only in some permanent molars.

1.2.2.2 Translocation 21:

Translocation occurs in about 3-4% of people with DS. In this type, an extra part of chromosome 21 is attached or translocated on to another chromosome. An example is attachment of a part of chromosome 21 on chromosome 14 or 22, or translocation of a part

of chromosome 21 on a different location of the same chromosome 21.

1.2.2.3 Mosaic 21:

It is the third type of DS. Individuals with this type have extra chromosome 21 material in only some of the cells of their body but not all. The other cells have the usual pair of chromosome 21. One to 2 percent of people with DS have this type. Individuals with mosaic 21 often have less of the prominent physical features associated with DS and develop and function closer to the normal life expected for their normal agemates[9].

1.2.3 Physical characteristics of Down syndrome:

DS is characterized by many unique physical manifestations. The eyes slant upward and outward, and have an extra-thick fold of skin on the inner side of the eye. The face is flat, with the rest of the head smaller than average. In relationship to the rest of the body, the legs and arms are shorter, with the feet and hands rather flat and broad. A single crease is often found stretching across the palm of the hand, and the fifth finger curves in toward the other fingers [8].

1.2.4 General health in patients with Down syndrome:

While there was an evidence in the past pointing to a shortened life expectancy, it is clear that, thanks to the better medical care now provided for them, children with DS nowadays are living longer than in the past. One study, carried out in the United States, showed an average life span of 49 years as opposed to 25 years of age before 1980, and approximately 10 years of age in 1929[10].

In the past, congenital heart disease, leukemias, untreated hypothyroidism and infections had contributed well to that high mortality rate, whereas today, the causes of death are more likely to be complications of Alzheimer's disease. It remains to be seen what their ultimate life expectancy will be, but it seems unlikely to approach the national average mainly because of the burden of the neuro-degenerative disorders to which they are predisposed[11].

Individuals with DS are more susceptible to systemic health problems such as:

1.2.4.1 Congenital heart problems:

Septal defects (openings between the chambers of the heart) and mitral valve prolapse (the mitral valve falls backwards into the

left atrium causing a backward flow of blood through the valve) are common [12,13].

1.2.4.2 Vision problems:

Refractive errors causing myopia (near-sightedness), hyperopia (far-sightedness), or stigmatism (blurring of both close-up and far-away visions) are common. Cataract may also be present [13].

1.2.4.3 Auditory problems:

Fluid accumulation in the middle ear frequently seen in individuals with DS mayresult in mild to moderate hearing loss. Middle ear infections are also quite common[14,15].

1.2.4.4 Seizure disorders:

Ten percent of individuals with DS have seizure disorders[16]. Most respond favorably to anticonvulsant medication [17].

1.2.4.5 Alzheimer's disease:

Alzheimer's disease is another common finding in persons with DS, and is the most significant health risk for those older than 40. Alzheimer's disease is a progressive and degenerative disease of the brain, causing the loss of millions of nerve cells and the

appearance of senile plaques on the brain. Nearly all persons with DS who die after age 40 show evidence of Alzheimer's disease[18].

1.2.4.6 Endocrinal problems:

Individuals with DS are at increased risk for dysfunction of the thyroid gland, an organ which helps control metabolism.Hypothyroidism is most common, occurring in almost one third of those with DS. This can becongenitalor due to attack on the thyroid by the immune system[19].

Reproduction is also affected by DS. Women with DS are less fertile and often have difficulties such asmiscarriage, premature birth, and difficult labor. However, these women are capable of having children with normal complements of chromosomes[20]. Men with DS are almost uniformly infertile, exhibiting defects in spermatogenesis[21].

1.2.4.7 Hematological problems:

Haematological abnormalities are common in children with DS. They are mostly benign. Neonatal changes include polycythaemia, neutrophilia and thrombocytopenia. In later childhood changes include secondary polycythaemia, red-cell macrocytosis, increased red-cell distribution width, leukopenia, and

immune dysfunction. Transient myeloproliferative disorder occurs in about 5% putting this group at particularly high risk of subsequent leukaemia. A full blood count should be checked in all neonates with Down syndrome. Those with transient myeloproliferative disorder should be discussed with a paediatrichaematologist. Of all children with Down syndrome, 1–2% will develop acute leukaemia. Children with Down syndrome and acute myeloid leukaemia usually respond well to chemotherapy. Acute lymphoblastic leukaemia responds less well, and particular care must be taken to minimize risk of infection[22]. In contrast to hematologic malignancies, solid tumor malignancies are less common in DS, possibly due to increased numbers of tumor suppressor genes contained in the extra genetic material[23].

1.2.4.8 Gastrointestinal problems:

Down syndrome increases the risk of Hirschsprung's disease, in which the nerve cells that control the function of parts of the colon are not present[24].This results in severe constipation. Other congenital anomalies occurring more frequently in DS include duodenal atresia, annular pancreas, and imperforate anus. Gastro-

esophageal reflux disease and celiac disease are also more common among people with DS [25]

1.2.4.9 Growth disorders:

Growth parameters such as height, weight, and head circumference are smaller in children with DS than with individuals of the same age. Adults with DS tend to have short stature. The average height for men is 5 feet 1 inch (157 cm) and for women is 4 feet 9 inches (144 cm) [26].

Individuals with DS are also at increased risk for obesity as they age[27].

1.2.4.10 Axial skeleton disorders:

Persons with DS are at higher than normal risk for atlanto-axial instability, probably due to ligament laxity. Periodic screening, with cervical x-rays, is recommended to identify this abnormality[28].

1.2.4.11 Mental retardation:

Mental retardation is the main feature of DS and accounts for 17 percent of all mentally retarded cases in USA[29].

The word “mental retardation" implies slow intellectual development, while "mentally handicapped" implies that a child has

difficulties in understanding and learning, both are associated with Down syndrome. Yet, it is important to realize that progress in learning and development, is possible. The range in development and learning has been categorized by mental ability and measured by the intelligence quotient (IQ). Any individual with an IQ score of 75 or less, (the average being 100) is likely to be classified as mentally handicapped or retarded [30]. Individuals with Down syndrome usually have IQs between 25 and 50 [12].

1.2.5 Dental anomalies in children with DS in different populations:

Bell J.*etal*.in South Australia observed the prevalence of taurodontism in extracted lower molars of 12 out of 33 (36.4%) individuals with DS. It was suggested that the high prevalence of taurodontism was associated with a delayed ingrowth and fusion of the epithelial flaps of the developing root sheath and that this anomaly represented one of several characteristic morphological dental features in DS that result from decreased mitotic activity of cells in developing tooth germs [31].

Russell B. G. andKjaer I.**(**1994) studied the frequency and pattern of tooth agenesis in a Danish p**o**pulation with

Downsyndrome. The control group consisted of a normal Danish population. They found that individuals with DS had an occurrence of agenesis of teeth that was some 10 times greater than in the general population with a higher frequency in males (90.7%) than in females (69.8%).Agenesis occurred more frequently in the mandible than in the maxilla and most often on the left side. In this study, the highly significant differences were primarily found in the occurrence of agenesis of the mandibular central incisors, followed by the maxillary lateral incisors, the maxillary second premolars and the mandibular second premolars. They suggested that the dentition, with its many different anomalies, could be used as an indicator in evaluating different aspects in the pathogenecity of aneuploidy conditions[32-34].

Cornejo*et al.* **(**1996) studied the oral health condition of Argentinean children and youngsters with DS in a sample of 86 Mongolic subjects aged 3 to 19, both sexes, resident in the city of Córdoba (Argentina), and compared it with a control group. The sample subjects were attending special educational institutions for the care of that type of disabled individuals. This population exhibited a high frequency of retarded eruption, agenesis,

conoidism, Angle's type III malocclusion, posterior cross bite and deficient gingival health[35].

Concomitant hypodontia and supernumeraries in DS cases were reported to be relatively rare. Only one case was reported in the literature[36].

Satsuki Kumasaka*et al*. (1997) used clinical records and panoramic radiographs to investigate developmentally absent permanent teeth in 98 Japanese subjects with DS. This retrospective study was based on the records and panoramic radiographs of subjects from approximately five years of age through to their most recent records. The time period covered ranged from 6 to 28 years. The majority of subjects with DS (63%) exhibited oligodontia, and many subjects were missing two or more teeth (53%) [37].

In Turkey (1997) 22 patients with DS were studied to determine the frequency of taurodontism. Eruption delay and congenitally lost teeth were also evaluated. While no taurodontic teeth were found in a control group of the same age, the frequency percentage of taurodontism was found as high as 66% ($p < 0.01$, Mann-Whitney test) [38] .

Rajic Z, Mestrovic SR (1998) in Zagreb examined 43 pictograms of subjects with DS. The grade of taurodontism, sex distribution and individual tooth involvement were recorded. Taurodontic teeth were found in 55.8% of the subjects, 32.6% females and 23.2% males. Second molars were most frequently affected (53.2%), followed by first molars (40%) and third molars (6.5%)[39].

There are few reports about the proportion of tooth wear in DS children worldwide. An investigation conducted by Bell E. J.*et al*.in Australia 2002 compared the aetiology, prevalence and severity of tooth wear in 49 cytogenetically confirmed Down syndrome children with 49 non-Down syndrome controls. In this study tooth wear was significantly more frequent ($p<0.01$) in the Down syndrome than the non-Down syndrome sample, with more of the Down syndrome children showing severe to very severe wear [40].

Shapira*et al*. studied dental anomalies in 34 individuals with Down syndrome in Israel. Excluding third molars, 59% of the individuals had missing teeth and 25% had small or peg-shaped upper lateral incisors [41, 42].

Bhowate R, Dubey A. studied 27 Indian DS children aged 10-14. They reported microdontia in 40.7% of their sample, while delayed eruption was reported in 14.8% [43].

Moraes *et al.* (2007) evaluated the incidence of dental anomalies in Brazilian patients with DS. A sample of 49 panoramic x-rays of DS patients aged 3 to 33 years was used. They divided the anodontia in two types: proven anodontia and suspected anodontia. Proven anodontia was considered when there was radiographic confirmation of the presence of a primary tooth and absence of its permanent successor tooth germ. While absence of the primary predecessor tooth with absence of the permanent tooth was considered as suspected anodontia. Proven anodontia was found in 17 individuals with 44 involved teeth, representing 34.69% of the sample. The cases with suspected anodontia occurred in 9 subjects with 19 involved teeth, representing 18.36% of the sample. Most cases of suspected anodontia occurred with maxillary lateral incisors. The characteristics of dental anomalies were observed in the panoramic radiographs in both the primary and permanent dentition, according to the ICD (International Classification of Diseases). There was a high incidenceof different types of

anomalies, such as taurodontism (50%), proven anodontia (20.2%), suspected anodontia (10.7%), conic teeth (8.3%) and impacted teeth (5.9%) [6].

Folakemi O. conducted a study to determine the oral health condition and treatment needs of a group of individuals aged 6-20 years old with DS in Nigeria. Participants were examined for oral hygiene status, dental caries, malocclusion, hypoplasia, missing teeth, crowding and treatment needs. Findings were compared with controls across age group, sex and educational background of parents. The results showed that participants with DS had poorer oral hygiene than controls, with no significant sex difference. Oral hygiene was similar in the lower age groups but deteriorated with age in the DS group. Missing teeth and malocclusion were recorded [44].

Sharath A. *et al.* (2008)studied the common oral findings and anomalies of DS children in Chennai city, India. Among the 130 DS children examined, 102 children aged 15 years and below were included in the study. A specially prepared clinical examination form was used to record the following findings in each child: a brief family and personal history; anomalies of soft tissues, teeth,

occlusion, and temporomandibular joint. Age-wise and gender-wise comparisons of the findings were done**.** About 97 children (95%) had the habit of regular tooth brushing. Everted lower lip (66%), retained primary teeth (31%), and midface deficiency (76%) were the most commonly seen soft tissue, dental, and occlusion anomalies, respectively [1].

Ingalls and Butler reported that the maxillary lateral incisor was congenitally absent in 25% of the children with Down syndrome[45].

A previous study studied the prevalence of anodontia in theprimary and permanent dentitions. The percentages of affected permanent teeth were: maxillary lateral incisor (31%), maxillary premolars (13%), mandibular central incisor (11%), mandibular lateral incisor (8%) and mandibular premolars (8%)[46].

1. 3 Justification of the Study

Children with DS are known to have many dental anomalies which might lead to dental problems. The mental capacity and the complex medical condition of these children may be a barrier for them to receive regular dental care; hence they need a specially organized dental health care program. Obtaining accurate data about this category of patients helps in reaching this goal.

There is no previous data available about dental anomalies among Sudanese children with DS.

1.4 Objectives of the Study

1.4.1General objective:

The general objective of this study was to assess the dental anomalies among a group of institutionalized Sudanese children with DS aged 6 to 14 years.

1.4.2 Specific objectives:

- To identify dental anomalies related to tooth number, shape, size, and eruption among DS children in Khartoum State.
- To determine the proportions of these anomalies and their distribution according to gender.

Chapter Two

Materials and Methods

2.1 Study Design:

This study was a cross-sectional, descriptive, institution-based study.

2.1.1Study area:

For proper clinical dental examination and for the availability of DPTs children with DS transferred from their centers to the clinic of the Division of Pediatric Dentistry, Faculty of Dentistry, U of K.

2.1.2 Study population:

Sudanese children (determined from their records) aged (6-14) years with DS attending the Rehabilitation Centers of Children with Special Needs.

2.2 The Criteria for Selection

2.2.1 Inclusion criteria:

Sudanese childrenwith DS who were:

- Attendees to rehabilitation centers
- aged 6-14 years

2.2.2 Exclusion criteria:

- Non-institutionalized childrenwith DS.
- Age group less than 6 years and more than 14 years.
- Hospitalized DS children with severely debilitating diseases.
- Definitely negative children according to Franklbehaviour rating scale.

2.3 Sampling:

The basic design was through simple randomization.

Total number of institutionalized DS children in Khartoum State was: 236

Total sample size was: 92

This was calculated according to the equations:

N (sample)= FN

$N=Z^2 1\text{-}a/2*P*(1-P)/(C\ \%)^2$

Where:

a= 0.05

P=0.5 (expected population)

c=Confidence Interval length.

F=(population size) /(population size)+N

Sample size was:

N (sample)= FN

$N=1.96^2*0.5^2/(0.08)^2 = 150$

$F=236/150+236 =0.611$

Sample size= 0611× 150 =92

2.4Ethical Considerations:

The proposal for this research was submitted to the Research Board, Faculty of Dentistry, University of Khartoum and after its approval, parents or guardian were told verbally, and then signed a written informed consent for the clinical and radiographic examinations and all necessary approval letters were obtained prior to conduction of the study.

The results of this study will be used where needed for the public without any violation of participants' privilege of secrecy. (see Appendices).

2.5 Data Collection and Management

2.5.1 Study tools and techniques:

A structured data collection form was developed, (Appendix 1). The data were collected by the same researcher conducting the study .The dental anomalies in DS children were evaluated by**:**

2.5.1.1 Clinical dental examination:

The clinical examination was conducted in the clinic of Pediatric Dentistry,U of K, on a dental chair under artificial light using a dental mirror and a ball-tip probe. The participants were examined for the presence of anomalies such as missing and extra teeth, abnormal tooth shape/size and delayed tooth eruption or any other dental anomaly such as over retained primary teeth and tooth wear. Only the presence or absence of anomalies was recorded. The severity of involvement was not recorded.

2.5.1.2 Radiographic examination:

Standardized dental panoramic tomographies(DPTs) were taken for all the DS patients in the Radiographic Diagnostic Clinic, Faculty of Dentistry. The DPTs were interpretated by the same investigator, under supervision. The cases and DPTs were categorized into two categories: those with and those without dental anomalies.

DPTs with dental anomalies were further studied to confirm the clinical findings in situations of missing teeth, extra teeth and abnormal tooth shape/size for both the primary & permanent dentitions. When retained primary teeth were recorded clinically,

the DPT was read to investigate the condition of the permanent successor for delayed eruption or agenesis.

2.5.2Data analysis:

The data were organized and entered in spread sheet using software SPSS (Statistical Package for Social Science) version 17 for statistical analysis.

2.6 Funding:

This research was self funded.

Chapter Three

Results

The sample size of Sudanese children with Down syndrome included in the study was 92. Their mean age was 10.4 (± 2.8) years. Their age characteristics are shown in Table 1.

Table 1: Age characteristics of the sample of Sudanese children withDS (in years):

Mean age	**10.4**
Median	10
SD	**2.8**
Minimum	6
Maximum	**14.2**

The sample was homogenous with respect to gender. Distribution of the study population according to gender is shown in Table 2.

Table 2: Distribution of the DS children according to gender:

	Frequency	**Percentage**
Male	43	46.7%
Female	49	53.3%
Total	**92**	**100%**

DS children were categorized into two groups; those without, and those with dental anomalies. Thedistribution of children according to the presence or absence of dental anomalies is shown in Table 3.

Table3: Distribution of DS children according to the presence or absence of dental anomalies:

	Frequency	**Percentage**
Children without dental anomalies	41	44.6%
Children with dental anomalies	51	55.4%
Total	**92**	**100%**

The group of DS children without dental anomalies was not studied further. The group with dental anomalies was then analyzed in details.

The presence of dental anomalies was found to be evenly distributed in the syndromicpatients as far as gender is concerned. This is shown in Table 4.

Table 4:Distribution of DS children with dental anomalies according to gender:

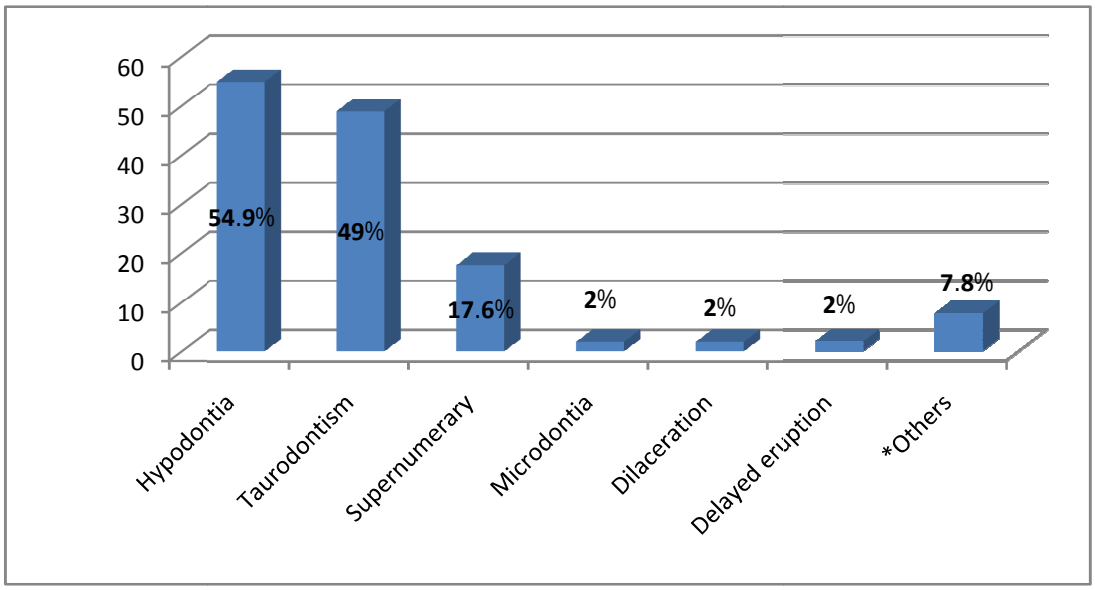
	Frequency	**Percentage**
Male	26	51%
Female	25	49%
Total	**51**	**100%**

Hypodontia was the most common dental anomaly found in this group of Sudanese DS children when the specific types of dental anomalies were analyzed. Its frequency was higher whether seen as a single dental anomaly, or as part of multiple anomalies occurring simultaneously in the same child.

Taurodontism came as the second most common anomaly, followed by supernumerary teeth.

The different types of dental anomalies in children and their frequencies are shown in Figure 1.

Figure 1: Frequencies of different types of dental anomalies in DSchildren:



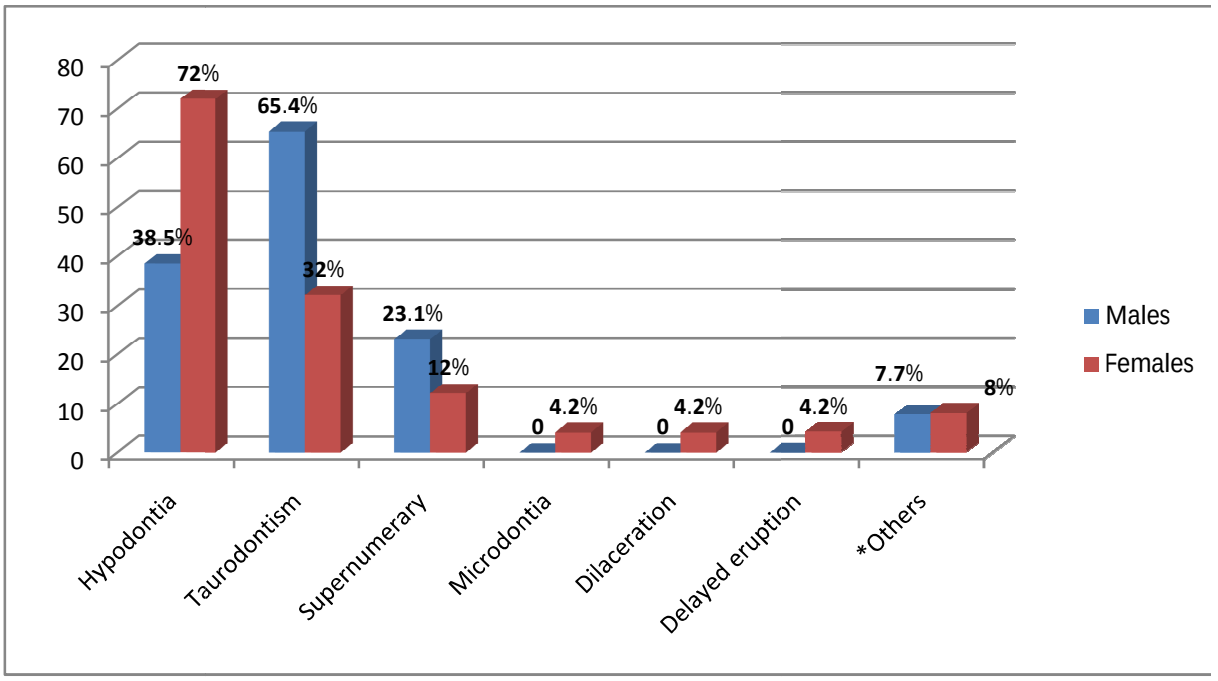
*others: Three cases of retained deciduous and one case of tooth wear.

The sum of the percentages in Figure 1 surpasses 100% because the percentages referred to the incidence of each anomaly in relation to the total sample size and several patients had more than one type of anomaly.

When these anomalies were studied according to gender, it was found that hypodontia was much higher in females while taurodontism was much higher in males. The differences in both findings were statistically significant. The P value was 0.016 and

0.017 respectively (Chi^2 test performed, P value <0.05 is significant). The details of gender distribution of anomalies are shown in Figure 2.

Figure2: Distribution of dental anomalies according to gender



*others: Three cases of retained deciduous and one case of tooth wear.

It was also found that almost two-thirds of the syndromic children had only one type of anomalies. Multiple anomalies in the same child, however, were extremely uncommon. The distribution of children according to the number of dental anomalies is shown in Table 5.

Table 5: Distribution of children according to the number of dental anomalies:

	Frequency	Percent
One anomaly	34	66.7%
Two anomalies	16	31.4%
Three anomalies	1	2.0%
Total	**51**	**100%**

The distribution of children according to the specific types of anomalies is shown in Table 6.

Table 6: Distribution of children according to the specific types of dental anomalies:

Anomaly		**Frequency**	**%**
<u>One anomaly:</u>			
	Hypodontia	18	52.9
	Taurodontism	9	26.5
	Supernumerary	4	11.8
	Others	2	5.9
	Microdontia	1	2.9
	Total	**34**	**100**
<u>Two anomalies:</u>			
	Hypodontia & taurodontism	8	50
	Supernumerary & taurodontism	4	25
	Taurodontism & others	2	12.5
	Hypodontia & supernumerary	1	6.25
	Taurodontism &dilacerations	1	6.25
	Total	**16**	**100**
<u>Three anomalies</u>			
	Hypodontia, taurodontism & delayed eruption	**1**	**100**

Distribution of the most common anomalies in different permanent teeth:

1. **Hypodontia:**

Hypodontia was found to be more common in the mandible than in the maxilla (41.3% and 58.7% respectively). It was more frequent in the mandibular second premolar area (mean percentage was 34.7%) followed by the mandibular lateral incisors (21.4%) and the maxillary lateral incisor area (17.3%). The distribution of hypodontia in the permanent teeth is shown inTable7**.**

Table 7: Distribution of hypodontia in different permanent teeth:

Tooth #	**17**	**16**	**15**	**14**	**13**	**12**	**11**	**21**	**22**	**23**	**24**	**25**	**26**	**27**	**Total**
Percentage	**2.7**	**0**	**5.3**	**4**	**0**	**9.3**	**0**	**0**	**8**	**0**	**4**	**5.3**	**0**	**2.7**	**41.3%**
Tooth #	**47**	**46**	**45**	**44**	**43**	**42**	**41**	**31**	**32**	**33**	**34**	**35**	**36**	**37**	**58.7%**
Percentage	**0**	**0**	**16**	**1.3**	**0**	**10.7**	**0**	**0**	**10.7**	**0**	**1.3**	**18.7**	**0**	**0**	
Total															**100%**

2. Taurodontism:

Taurodontism, the second most common anomaly,was found only in some permanent molars. All premolars and anterior teeth had normal morphology as far as taurodontism was concerned.Its distribution is shown in Table 8.

Table8: Distribution of taurodontism in different permanent molars:

Tooth #	**17**	**16**	**26**	**27**	**Total %**
Percentage	**9.8**	**23**	**18**	**1.6**	**52.5**
Tooth #	**47**	**46**	**36**	**37**	
Percentage	**0**	**21.3**	**26.2**	**0**	**47.5**
Total					**100%**

Supernumerary were seen in 9 cases; 6 were males & 3 were females. Most of them seen adjacent to tooth #12 followed by #13, #14, # 42 and #22.

Only one case of microdontia was recorded; a male, all of his teeth were affected. Similarly, only one case of dilaceration and

another case of delayed eruption were recorded, all of them were seen in females.

Three cases of retained deciduous teeth were seen; two of them were in females while one was in a male. Only one case of tooth wear was seen in a male.

Chapter Four

Discussion

Epidemiological studies on dental anomalies in children with Down syndrome are scarce, and those few reported ones are not quite comparable due to wider range of age, fewer numbers of cases in each study and may be different research methodologies.

In the rehabilitation centers where the sample of DS children was randomly selected, children younger than 6 years of age were not available. The mean age was about 10 years of age. The sample was homogenous with respect to gender.

In this group of Sudanese children with DS, a high proportionof dental anomalies was found reaching 55.4%.

About two-third of the DS children (66.7%) had one anomaly and one-third (31.4%) had two. Only one case of DS had three anomalies. These findings are different from a similar study in Brazilian DS children in whom, although the proportion of dental anomalies was also high, but in most cases the same individual had more than one dental anomaly.

In our study, hypodontia (a term used here for the same meaning as agenesis or anodontia) was the most commonly observed dental anomaly, affecting 54.9% of DS cases.

Moraes *et al.*conducted a retrograde study in which only DPTs of Down syndrome cases were studied. Because age variation of tooth development in general, and in DS patients in particular, they divided the anodontia in two types: proven anodontia and suspected anodontia. In that study, proven anodontia was considered when there was radiographic confirmation of the presence of a primary tooth and absence of its successor tooth germ, while absence of the primary predecessor tooth with absence of the permanent tooth was considered as suspected anodontia. In our opinion, the suspected hypodontia should be when it is not quite sure that the permanent tooth is congenitally absent, for a permanent tooth might develop very late. Proven anodontia in that study represented 34.69% of the sample and suspected anodontia represented 18.36% of the sample. Most cases of suspected anodontia occurred with maxillary lateral incisors [6]. The proportion of hypodontia seen in our study is higher than what was observed by Moraes *et al.* although only proven hypodontia was

recorded, suspected hypodontia was considered normal. This might have underestimated the actual number of cases with hypodontia in this group of Sudanese children with DS. Hypodontia in these children however, is lower than what was reported by Kumasaka *et al.* who found that 63% of his sample had hypodontia [37].

Our results are similar to Folakemi O. who found more than half of the Down syndrome group had one or more missing teeth [44].

Shapira*et al.*[41-42]and Cornejo*et al.*[35] have also reported a high frequency of agenesis among Israeli and Argentinean children with DS respectively, similar to what we have observed in this study.

Females were more affected than males. This contrasts a study among Danish DS children in whom agenesis was more frequent in males than in females[32-34].

The finding that hypodontia occurred more frequently in the mandible than in the maxilla in our study, coincides with what was found among Danish DS children in whom agenesis occurred more frequently in the mandible than in the maxilla[32-34].

Hypodontia was more frequent in the mandibular second premolar area (34.7% in both right and left sides) followed by the mandibular lateral incisors (21.4%) and the maxillary lateral incisor area (17.3%). These findings contrast the findings of a study of the prevalence of anodontia in the primary and permanent dentitions, where the percentages of affected permanent teeth were: maxillary lateral incisor (31%), maxillary premolars (13%), mandibular central incisor (11%), mandibular lateral incisor (8%) and mandibular premolars (8%)[46]and also Kumasaka *et al.*[37]who reported that the most frequently absent tooth was the mandibular lateral incisor (23.3%) followed by maxillary second premolars (18.2%), maxillary lateral incisors (16.5%) and the mandibular second premolars (15.3%).

The teeth most frequently missing in Folakemi O. study were the maxillary lateral incisors, followed by mandibular lateral incisors[44]. This is different from the finding from our study mentioned above.

The percentage of congenitally absent upper laterals seen in this study, however, is lower than what was reported by Ingalls and

Butler who reported that the maxillary lateral incisor was congenitally absent in 25% of the children with DS [45].

Taurodontism was the second dental anomaly observed in DS Sudanese children in this study, being identified in 49% of the DS individuals. It was also noted that taurodontism affected several teeth of the same patient and occurred mostly in the same tooth type (permanent molars only).

The prevalence of taurodontism in this study is different from Moraes *et al.*[6] who found a higher incidence of taurodontism (85%).This higher percentage may be because of the fewer number of DS cases in their study compared to ours. However, the proportion of children with taurodontism in our study is higher than the study done by Bell J. in South Australia who observed taurodontism in extracted lower molars of 36.4% individuals with DS [31], but lower thanAlpoz and Eronat who reported a frequency percentage of taurodontism in 66% of individuals with DS [38]. The variations in the prevalence of taurodontism reported in these studies can mostly be related to the variation in the age groups examined.

Rajic and Mestrovic [39] in Zagreb found taurodontic teeth in 55.8% of the subjects. Their results however, are different from our findings as far as gender and tooth type are concerned; in their studies females are more affected than males (32.6% and 23.2% respectively), while in our study males suffered more from taurodontism than females, (65.4% and 32% respectively). Also in their study the second permanent molars were the most frequently affected teeth (53.2%), followed by the first molars (40%). This contrasts our present findings in which the first permanent molars were more frequently affected (47.5% of the lower molars and 41% of the upper molars) than the second permanent molars (in which only the upper ones were affected with a percentage of 11.4 while the lower second molars were not affected).

Although supernumerary teeth were found in 17.6% of our sample, concomitant hypodontia and supernumeraries were observed in only one case. This is similar to what was reported in the literature [36].

Other dental anomalies were observed in sporadic cases ; only one case of root dilaceration involving two teeth (#35 & #15) in a same patient, a case of microdontia; and a case of delayed tooth

formation and eruption, each one of them representing 2.0% of the sample, a finding very similar to Moraes findings[6].Shapira however, reported a very high incidence (25%) of microdont upper lateral incisors among Israeli DS children [41,42].

Scully stated that the eruption of primary and permanent teeth was delayed in individuals with DS and that the primary teeth not always had their formation completed before the age of 5, and that alterations in the eruption sequence might occur[47]. Likewise, Coelho and Loevy reported delayed tooth formation and eruption, alteration in the eruption sequence as well as microdontia and anomalies of shape[46]. Our study has failed to confirm these findings; very few cases of delayed tooth eruption were observed. Retained deciduous and tooth wear (designated in this study as "others") were also uncommon among our sample, a finding different from Sharath Asokan, *et al.*[1] who found that retained primary teeth were the most common dental anomaly present in his study (31%). However, unlike our findings, tooth wear was a significant finding in Australian DS children [40].

The outcome of the present study is similar to those of the reviewed literature with respect to the most common types of

anomalies identified in DS; except for hypodontia and taurodontism, other anomalies showed a low incidence.

Limitation of the study:

- In this group of Sudanese children with DS, only dental anomalies were studied, i.e. those developmental anomalies affecting the hard dental tissues. Dental caries and malocclusion were not investigated. Further studies to assess these conditions as well as soft tissue anomaliesare recommended.
- A second limitation in this study is that, only institutionalized DS children were studied, non-institutionalized at-home children could not be reached.

Conclusion:

- The prevalence of dental anomalies in Sudanese DS children found in the present study is similar to those of the reviewed literature with respect to the most common types of anomalies identified in Down syndrome.
- Except for hypodontia and taurodontism which were quite common in our sample, other anomalies showed a low incidence.
- When these anomalies were studied according to gender, it was found that hypodontia was much higher in females while taurodontism was much higher in males.
- Dental anomalies in Down syndrome children may exhibit great variability as seen when comparing the reported findings in the literature within themselves and also when we compare our findings to these reports.

Recommendations:

- Children withDown syndrome belong to those groups of children with special needs. Their medical condition reinforces that the need for preventive as well as early conservative dental care should be given a high priority to prevent and/or control problems that may affect them, for example the need for endodontic treatment in taurodontic teeth, a highly incident dental anomaly in these children.
- In the present study, only the presence or absence of dental anomalies was considered, leaving the analysis of its severity for future investigations.
- Further research is highly recommended to collect baseline data on the oral health of this group of children with special needs, upon which preventive as well as early interceptive dental treatment strategies can be built up.
- The first dental appointment for the child with Down syndrome should be at one year of age.
- At this appointment, the dental staff should examine the child and discuss the etiology of dental diseases, how to prevent them and the consequences of untreated diseases (tooth loss)

with the caregiver; as most of these children already suffer from hypodontia.

- Ø Individuals with Down syndrome learn better with visual teaching, so pictures, models, and diagrams are an important part of the teaching and learning process.
- Ø Instructions should be given slowly, or one concept at a time. To insure understanding, the child should demonstrate brushing and flossing techniques. Down syndrome cases are generally motivated to please. Therefore, positive verbal reinforcement should be used generously.
- Ø Supervision of toothbrushing may still be necessary in those with partial independency to ensure that the child's efforts are effective.
- Ø Fluoride use-in Sudan mostly in the form of fluoridated tooth pastes- should be stressed in daily basis.
- Ø Ministry of Health in Sudan is respectfully requested to investigate the prevalence of Down syndrome among Sudanese children. If this information would be available, then the prevalence of oral diseases in general and dental anomalies in particular, could be investigated in more

detailed, hence preventive measures could be tailored to this group of children.

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Appendix (I)

جامعة الخرطوم

كلية طب الأسنان

مجلس أبحاث الكلية

قسم التقويم و طب أسنان الأطفال و طب الأسنان الوقائي

شعبة أسنان الأطفال

Data collection sheet

Personal data

Name of the Rehabilitation Centre……

Patient's codes:……..

Date of birth ……Age ..… yrs …..mon

Gender: ……

Phone No: ……

Medical health ……

Taking any medications?

No …. Yes ….. (Specify) ……..……..

Diagnosis according to the medical

records:……..……

……

Clinical examination:

Patient has dental anomaly?

No.....Yes…… **If Yes,**

Type of dental anomaly	**Tooth #**
Hypodontia	
Supernumerary	**Adjacent to tooth #**
Microdontia	
Macrodontia	
Gemination	
Fusion	
Concrescence	
Taurodontism	
Dilaceration	
Delayed eruption	
Others	

Radiographic examination & interpretation of the DPTs:

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Appendix (2)

جامعة الخرطوم

كلية طب الأسنان

مجلس أبحاث الكلية

قسم التقويم و طب أسنان الأطفال و طب الأسنان الوقائي

شعبة أسنان الأطفال

التاريخ / / 2010

السيدة /الدكتورة نادية أحمد يحيى/ عميد كلية طب الأسنان/رئيس مجلس أبحاث الكلية .....

المحترمة

السيدة/الدكتورة فاطمة الخضر الحسن /مقرر درجة ماجستير طب أسنان الأطفال .....

المحترمة

السلام عليكم و رحمة الله تعالى و بركاته

الموضوع /طلب موافقة بإجراء بحث أطروحة لنيل درجة الماجستير في طب أسنان الأطفال

أتقدم لسيادتكم بطلب للموافقة على إجراء بحث أطروحة درجة الماجستير في طب أسنان الأطفال،

بعنوان :-

Dental Anomalies among Down Syndrome Children in Khartoum State(Sudan).

علماً بأن البحث يعتمد على فحص سني إكلينيكي روتيني يتم في عيادة الأطفال بالكلية، مع عمل صور شعاعية بقسم الأشعة بالكلية ، وسيتم أخذ موافقة كل المشتركين في البحث و أولياء أمورهم قبل البدء في عملية الفحص.

ولكم جزيل الشكر و التقدير

الدكتورة/ رقية حسن عثمان فرحات

طالبة الدراسات العليا لنيل درجة الماجستير في طب أسنان الأطفال

Appendix (3)

جامعة الخرطوم

كلية طب الأسنان

مجلس أبحاث الكلية

قسم التقويم و طب أسنان الأطفال و طب الأسنان الوقائي

شعبة أسنان الأطفال

التاريخ / / 2010

السيد/مدير

منظمة/مؤسسة.....المحترم

السلام عليكم ورحمة الله تعالى وبركاته

الموضوع / إجراء بحث طبي

بالإشارة للموضوع أعلاه، يجري قسم أسنان الأطفال بكلية طب الأسنان – جامعة الخرطوم بالتعاون مع مجلس الدراسات الطبية والصحية العليا بحثا لجمع معلومات عن الصحة الفموية لدى الأطفال ذوي الاحتياجات الخاصة في ولاية الخرطوم. وذلك من خلال فحص طبي روتيني وصور اشعاعية فقط دون إجراء أي عملية أو أخذ أي عينة وسوف يتم الفحص بقسم أسنان الأطفال بكلية طب الأسنان جامعة الخرطوم.

نأمل في كريم تفضلكم بالتكرم بالموافقة على إجراء هذا البحث في منظمتكم العامرة.

ولكم جزيل الشكر و التقدير

مقدمة الطلب د/ رقية حسن عثمان فرحات

طالبة دراسات عليا بجامعة الخرطوم، كلية طب الأسنان

اعتماد إدارة المنظمة/المؤسسة

التوقيع: .....

تلفون المنظمة/ المؤسسة .....

التاريخ: .....

Appendix (4)

جامعة الخرطوم

كلية طب الأسنان

مجلس أبحاث الكلية

قسم التقويم و طب أسنان الأطفال و طب الأسنان الوقائي

شعبة أسنان الأطفال

التاريخ / / 2010

استمارة موافقة للمشاركة في البحث

السيد/ ولي أمر الطفل :..... المحترم

السلام عليكم و رحمة الله تعالى و بركاته

يجري قسم أسنان الأطفال بكلية طب الأسنان، جامعة الخرطوم بالتعاون مع مجلس الدراسات الطبية والصحية العليا بحثاً لجمع معلومات عن الصحة الفموية لدى الأطفال من ذوي الاحتياجات الخاصة في ولاية الخرطوم . وذلك من خلال فحص سني روتيني وأخذ صور شعاعية بعيادة أسنان الأطفال بكلية طب الأسنان، جامعة الخرطوم، و قد وقع الأختيار على المنظمة / المؤسسة التي يدرس بها ابنكم/إبنتكم المذكور(ة) أعلاه ضمن نطاق العينة.

بناءً على ما تم ذكره، نرجو كريم موافقتكم الشخصية للكشف على ابنكم/ابنتكم. ونؤكد و نضمن لكم ألا تستخدم المعلومات المجموعة لغير الأغراض الدراسية.

أقر أنا المذكور أدناه بموافقتي على اشتراك ابني/ابنتي في البحث كمتطوع. علماً بأنني قد تفهمت أهداف البحث.

اسم ولي الأمر .....

رقم الموبايل .....

رقم تلفون المنزل .....

التوقيع .....

التاريخ .....

و الله الموفق